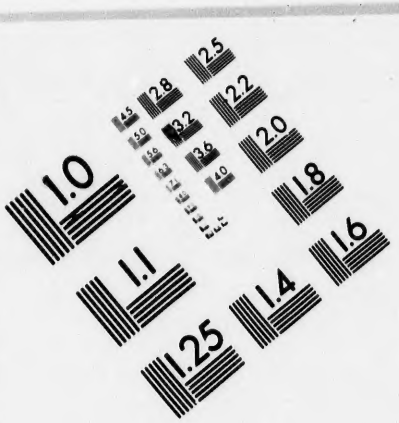

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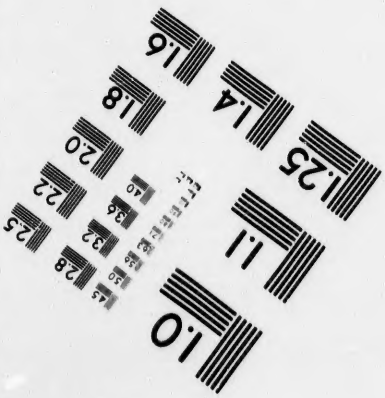

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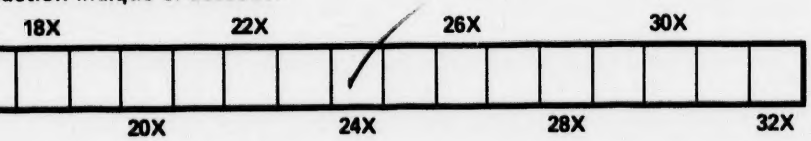

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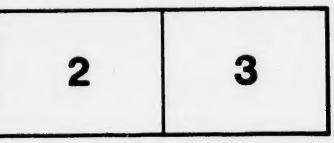

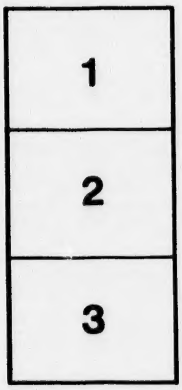

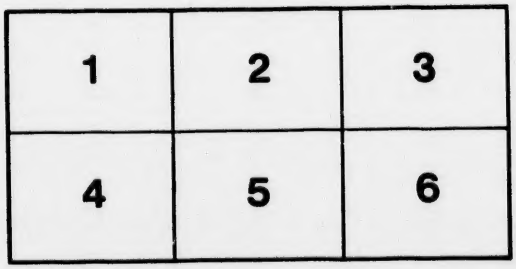

McGILL UNIVERSITY

PAPERS FROM THE DEPARTMENT

OF

PATHOLOGY.

No. 2.—Notes upon a Case of Madura Foot.
(Mycetome Pedis, Ochroid Variety.)

BY

J. G. Adami, M.D.,

AND

R. C. Kirkpatrick, M.D.

[Reprinted from the Transactions of the Association of American Physicians, 1895.]

Montreal, 1896.

A CASE OF MADURA FOOT DISEASE.

(Mycetoma Pedis, Ochroid Variety.)

By J. GEORGE ADAMI, M.A., M.D., M.R.C.S. Eng.,
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Interest in this curious disease, mycetoma pedis, has been stimulated during the last three or four years by the researches of Kanthack,[13, 14] and of Boyce and Surveyor[4, 5, 6, 7] in England, of Vincent in Algeria,[10, 18] and of Bassini [1, 16] in Italy; and, whereas, it used to be considered as essentially a local disease, occurring only in India, and mainly in the Madura district, and in the Province of Scinde, we now know that it has been met with elsewhere: in Italy, in Algeria, and (I have this on the authority of Dr. Kanthack) in Germany.

The case we record is, we believe, the first on this continent in which we are dealing with the true disease in a patient who has passed all his life in America. We say this with some hesitancy, for there are not a few features in connection with Kemper's case,[15] published nineteen years ago, which render it probable that the appearances described were those of true Madura foot. Nevertheless, in more than one particular the short account given introduces variations from the typical symptomatology. To these we shall recur later. Another case occurring in the practice of Dr. Charles T. Parkes, of Chicago (quoted by Shoemaker), is invalidated in this connection by the fact that the patient had resided for many years in India.

The following notes of this case were obtained by one of us (R. C. K.) with some little difficulty from the dense and taciturn patient.

Xavier Lacompte, a French-Canadian, aged twenty-one years, was born in Montreal, and has always lived there, save for five years, between the ages of

twelve and seventeen, passed in the Province of Ontario. He had never been out of Canada. His parents, three brothers, and four sisters are living and in perfect health. There is no history of tuberculosis in the family. When he was eleven years old a bluish spot appeared on the inner side of the right foot; this gradually increased in size until it was as large as a five-cent piece. One day, while walking, he struck the foot, breaking open the spot, and from it a little blood escaped; after this the spot disappeared. A few months later there appeared on the sole of the foot below the first and second metatarsals what the patient described as "un bouton de chair"—a button of flesh. After a time this was removed, leaving a little hole which ultimately healed. Three years later a similar growth appeared on the dorsum of the foot, directly opposite to where the other nodule had been on the sole. Two years later he struck the foot with an axe, bruising it severely; from that time it remained swollen and tender.

FIG. 1.

Apparently the patient never suffered severe pain, and he was able to use the foot and to walk until the autumn of 1893, when the condition became aggravated, button-like nodules developing all over the foot. As shown by the illustrations (Figs. 1 and 2), these buttons were in the main discrete, but situated in groups, of which the most extensive was upon the dorsum of the foot, from below the external malleolus downward along the outer side; another

group extended over the metatarsals of the hallux and second toe, passing into a group about the bases of the first three toes. There were other groups situated over the internal malleolus, in the instep and over what, for the sake of brevity, may be termed the pad of the second and third toes. The isolated buttons, and they were the majority, were of the size of a pea or somewhat larger. Here and there two or three such buttons were confluent. Where they were isolated they were sub-pedunculated, the circumference

FIG. 2.

beyond the base being greater than at the base. Each of them represented a cutaneous overgrowth of low vitality, pinkish or bluish-pink in color, formed around the opening of a sinus. Over the middle portion of the dorsum of the foot, and to a less extent elsewhere, small cicatrices were observable, due evidently to breaking down and healing with closure of the sinuses. These sinuses were very extensive; in fact the foot was riddled with them, and, as I found upon examination after removal of the foot, they passed deeply in all directions, and here and there led to carious bone.

The muscles of the leg were atrophied, so that the leg contrasted strongly with the foot, which, from the ankle to the offset of the toes, was greatly swollen. Between these two regions there had been extensive destruction of the ligaments, for the bones could be felt freely moving on one another. How extensively the bones themselves had become affected was demonstrated

by a mounted preparation of the bones of the foot. There was an extreme condition of rarefying osteitis of all the tarsal and metatarsal bones, with caries. The bone destruction was greatest in connection with the cuboid and external cuneiform. The articular surfaces of the tarso-metatarsal series of joints had entirely disappeared save those of the apposed internal cuneiform and first metatarsal. Toward the periphery of the affected region—on either side of the os calcis below, on the cutaneous surfaces of the scaphoid and internal cuneiform, and on the proximal halves of the four outer metatarsals—were delicate, fairly long radiating osteophytes. There was a second small area of advanced rarefying osteitis affecting the proximal ends of the three middle phalanges. The disturbance in this area tallied with the history given of the first external appearances of the typical mycetoma buttons, while the condition of the rarefied bone with its small cysts or pits hollowed out in it tallied wholly with the description and figures of the Indian disease given by Vandyke Carter.

The foot being absolutely useless, Dr. Kirkpatrick removed it in June, 1894, performing a Syme's operation at the junction of the middle and lower thirds of the leg. Recovery was complicated by an attack of pleurisy, but eventually the patient left the hospital with a well-healed stump.

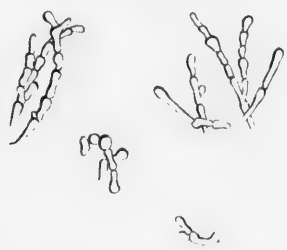
The specimen, we greatly regret to state, arrived at the Pathological Laboratory at McGill University at a time when one of us (A.) was preparing to depart for the summer, and in the stress of work was placed in preservation fluid before he had time to study it. When, two days later, he heard that it was to be brought before the Montreal Medico-Chirurgical Society, he examined it hastily, and was immediately struck by its resemblance to specimens of Madura foot disease. There were the same fleshy buttons with sinuses, and upon pressure upon them a thin pus exuded in which were granules of a yellowish-gray color, of various sizes up to that of a pin-head, and yet larger. None as large as a pea, was obtained, but some as large as two or three hempseeds moulded together. The larger masses were formed of conglomerations of smaller particles.

Upon studying the fresh discharge under the microscope, and again upon examination of sections, the structure of these bodies could be easily seen. They were identical in general appearance with actinomyces, forming lobate-reniform masses, with central dense mycelium and a radiate arrangement of filaments or clubs around the periphery. These clubs, however, were larger than those of actinomyces; in the fresh exudate they were easily distinguishable by the low power of the microscope (Zeiss A), while under the high power there was observable a marked tendency for the clubs to bifurcate. The masses were surrounded by leucocytes, and in many cases the collections were so loose that the fungi tended to fall out.

In the preserved specimens the clubs underwent considerable shrinking, so that they appeared not nearly so characteristic when seen *en masse*. Nevertheless in these, also, upon breaking up and teasing the masses, the branching could frequently be seen. Here and there in some of the masses long hyphal

processes tending to be swollen at their extremities projected far beyond the main clump of rays. In these, while there was evidence of granulation and uneven staining, I could detect no sign of transverse cleavage. In one specimen, however, I came across ample evidence of such, and it is necessary that the fact should mentioned. This was in a specimen of a large mass obtained from a sinus when first I examined the foot. This was placed in glycerin and surrounded with Canada balsam, and has formed one of the best and most typical examples of the actinomyces-like appearances that I have mounted. (Fig. 3.) Within the last few days I have teased and faintly stained a portion of this mass, and to my great surprise have found that some of the separated hyphæ show the clearest evidence of being formed of irregular joints of varying length and very varying breadth, some of the segments being oval, and, in fact, almost spherical, although in general they are elongated. Of such segmentation, as I have said, not a trace is observ-

Fig. 3.



able in the peripheral zone of the intact mass, and I am led to conclude either that these segmented hyphæ are an intrusion, that the sinus contained more than one form of fungus, or that in the deeper layers of the rayed masses true segmentation does exist, as Carter originally noted and figured, and as Bassini also found in his case.

Leaving out of account this single observation, I may say that in all other respects the microscopical appearances, in general, tally with the description given by Kanthack,[14] just as the clinical history and the appearance of the diseased foot tallied entirely with the description given by Vandyke Carter[8] and others.

There are two main varieties of mycetoma, the black and the white, or, more truly, ochroid. To these Vandyke Carter would add a third, in which the surface has the appearance of having been sprinkled with red pepper. So far as I can learn this third variety is very rare, and no exact observations have been made upon its pathology. It may, therefore, be passed over for the present.

*

Of the other two the black is much the more common in India. In this there is a deposit of blackish pigment within the mycelial masses. Whether we are dealing with a separate micro-organism, or whether the pigmentation is due to difference of age and of environment, is a matter that is not yet settled beyond dispute, although the clinical and other evidence is, taken altogether, somewhat in favor of the former supposition. The cases observed out of India have been all of the light-colored variety, with the one exception of Bassini's.

The further question as to whether this last form is or is not, as Carter suggested some years ago,[9] a local manifestation of actinomycosis, would seem to have been settled by Vincent's successful culture of the fungus obtained from one of his two Algerian cases.[18]

Turning now to this pale form or variety—what are its relationships? Is it or is it not a form of ray fungus, a variety of actinomyces? That it is not the ordinary actinomyces is evident from several considerations. While, occasionally, in the actinomycotic ox we may meet with the mycelial masses possessing giant rays, these are the exceptions, not the rule; only some of the club are gigantesque. Here, in mycetoma, all the peripheral clubs are large—much larger than those of the bovine actinomyces. Again, clinically, mycetoma is essentially a local disease. It may, as in our case, affect a foot for years, but it does not extend beyond or lead to a generalized morbid process with metastases.*

These two considerations alone are sufficient, it seems to me, to overthrow Vandyke Carter's suggestion that it is a local manifestation of actinomycosis. Were further proofs necessary we have Vincent's record of his successful cultivation of the fungus from one of his two Algerian cases. The pure growths obtained by Vincent, while being of the streptothrix type and so broadly resembling actinomyces hominis, presented so many points of difference, that if it be accepted that Vincent was dealing with an example of the true disease there can no longer be any doubt as to the difference between the two fungi.

The only difficulty in accepting Vincent's case as being one of the true disease lies in his diagram and description of the fungus *in situ*

* Not a little unnecessary confusion has been introduced in to the discussion of this subject by a misunderstanding of Kanthack's position. Kanthack, as his paper clearly shows, never attempted to prove that the madura fungus is identical with actinomyces hominis or bovis. only that it is an actinomyces, a ray fungus, belonging to the same group as the European ray fungus.

in the tissues. This does not correspond satisfactorily with the diagrams given by Kanthack and Hewlett, or with the appearances seen in our case. Instead of a peripheral zone of clubbed rays, the diagram shows fine broken filaments, some of which radiate outward for a long distance between the cells of the mycetoma tubercle. Yet Vincent gives the usual clinical history and symptoms, and his description of the appearance of the grains obtained from the discharge out of the sinuses is fairly well in accord with what we have noted in our specimens. It is possible, therefore, that Vincent has figured an atypical and far advanced mass rather than one that is typical.

To sum up, the descriptions given by various observers are so contradictory that the time is not yet ripe to make a positive statement as to the nature of the fungus of this disease; nor, unfortunately, do we make matters clearer by our case, with its microscopical characters in general confirming the view that it is allied to actinomyces, and nevertheless, in our specimen, the certain presence of segmentations of the filaments.

Lastly a few words require to be said upon the clinical history of the disease. As with actinomycosis, so here, local injury, such as the prick of a thorn, is the common history given of the origin of the disease.[2] Bocarro found the pad of areolar tissue along the bases of the toes to be the favorite initial seat of the lesions, and, as the names given imply, the foot is the organ in general affected. Its prevalence in connection with the foot is evidently due to the habit of walking barefooted. On this continent barefootedness is the exception; in India among the rural population it is the rule. It is interesting to note that the disease in the case here reported began in the age of barefootedness, namely, before the fourteenth year, and that the first pathognomonic development was upon the pad of the toes. Nevertheless other regions besides the feet may be primarily affected. Thus of Bocarro's hundred cases in the Province of Scinde,[2] three were upon the hand, one over the shoulder-blade, one in the region of the sacro-iliac joint. As to the age of incidence, the same authority gives the period between the twentieth and fortieth years as being that of most frequent development; but on referring to his table, the greatest number per decennium is found between the tenth and twentieth years.

In general the disease has a very chronic course; cases have been recorded of twenty-six and even thirty years' duration; the usual

duration is from three to seven years. In rare cases a considerable portion of the foot has become involved in twelve months or less.

Nevertheless if one case has occurred on this continent it is fairly certain that others have, and Kemper's case may have been one of these. I own that it is difficult to imagine what disease Kemper could have had before him if it was not mycetoma. It must, however, be remembered that recent observations are bringing to light the fact that aspergilli and other fungi have a tendency to assume a rayed growth within the organism.[6] The probability is that we have to deal with a series of actinomycoses, and it is a question which the future must solve, as to how many fungi, of the more usual type—not only streptothrices, but also hyphomycetes—are capable of producing the clinical appearances of Madura foot or mycetoma pedis. Bassini's case, for example, does not wholly conform with the classical descriptions of the black variety, nor again does Hewlett's third case.[12] We have pointed out that in some respects Vincent's case presents unusual features, and if further research in our own case brings to light additional examples of true segmentation of the hyphæ then it will have to be divided off from the rest.

In its slowly progressive character, its long duration, the absence of constitutional disturbance, and little local pain, in external appearance and the development of innumerable sinuses with affections of the bones of the foot, in the presence of characteristic large granules, recalling the appearance of enlarged actinomycotic masses—in all these particulars it will be seen that our case conforms remarkably with the disease as found in India. Kemper's case, on the other hand, presents many departures from the usual type. In it the patient, a native of Ohio, aged twenty-five years, suffered from a swollen foot for three months before any external lesion was manifest. Then several blebs the size of a split pea made their appearance. When first seen by Kemper there were five or six of these, about half an inch in diameter. In their centres a round opening existed with well-defined borders, and out of the sinuses exuded a glairy white fluid, resembling white of an egg and extremely offensive. These eventually coalesced into one large ulcer. Later other ulcers appeared. There was intense pain, so that the patient gained very little sleep. The foot was removed six months after the first swelling showed itself.

Examination after removal revealed a large ulcer over the inner

side of the foot, whose surface was depressed quarter of an inch below the general surface of the organ. This was covered by a white, friable, fluffy substance, appearing to the naked eye like mould or fungus. A probe could be passed into several sinuses opening into the ulcer. These sinuses contained masses of the same fluffy material as was found upon the surfaces of the ulcers, and, in addition, yellowish, highly refractile bodies, forming mulberry-like groups. The sinuses opened into cavernous spaces within the substance of the foot. These were filled with white tubercles.

It will be seen that this case of Kemper and Jameson varies from the usual clinical history and symptomatology in its short duration—six months—in the absence of any description of the very characteristic buttons, in the fact that redness, swelling, and tenderness of the foot preceded any recorded external inflammatory manifestation; and again in the fact that ulceration appears to have been the main process, and that the ulcers were covered with white, fluffy, mould-like substance. This last point together with the description of blebs rather than buttons are points which I think are unlike what would be described in connection with the true disease.

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